

Citation for published version:

Jones, A, Caes, L, Eccleston, C, Noel, M, Rugg, T & Jordan, A 2020, 'Loss-adjusting: Young people's constructions of a future living with Complex Regional Pain Syndrome', *The Clinical Journal of Pain*, vol. 36, no. 12, pp. 932-939. <https://doi.org/10.1097/AJP.0000000000000880>

DOI:

[10.1097/AJP.0000000000000880](https://doi.org/10.1097/AJP.0000000000000880)

Publication date:

2020

Document Version

Peer reviewed version

[Link to publication](#)

Publisher Rights

CC BY-NC

University of Bath

Alternative formats

If you require this document in an alternative format, please contact:
openaccess@bath.ac.uk

General rights

Copyright and moral rights for the publications made accessible in the public portal are retained by the authors and/or other copyright owners and it is a condition of accessing publications that users recognise and abide by the legal requirements associated with these rights.

Take down policy

If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

Loss-adjusting: Young people's constructions of a future living with Complex Regional Pain Syndrome.

Abigail Jones, MSc¹, Line Caes, PhD², Christopher Eccleston, PhD³, Melanie Noel, PhD⁴,
Tessa Rugg, MSc⁵, Abbie Jordan, PhD¹

¹Department of Psychology and Centre for Pain Research, University of Bath, UK. ²Division of Natural Sciences, University of Stirling, UK. ³Department for Health and Centre for Pain Research, University of Bath, UK. ⁴Department of Psychology, University of Calgary; Alberta Children's Hospital Research Institute; Hotchkiss Brain Institute, Canada.
⁵Department of Psychology, University of Bath, UK.

Corresponding Author: Abbie Jordan, PhD. Department of Psychology and Centre for Pain Research, University of Bath; Claverton Down, Bath BA2 7AY, UK; Telephone: 00 44 (0)1225-386982; Email: a.l.jordan@bath.ac.uk

Conflicts of Interest and Source of Funding: Dr Jordan and Dr Caes report a grant from Pain Relief Foundation to support the conduct of this study. Dr Jordan, Dr Caes, and Professor Eccleston report a grant from Rare Diseases Foundation to support the conduct of the study. The remaining authors have no conflicts to declare.

Acknowledgements: We would like to thank the Pain Relief Foundation and Rare Diseases Foundation for supporting this work. We would also like to thank the adolescents who participated in the study and CRPS organisations who supported the study through recruitment advertisements.

Abstract

Objectives: Complex Regional Pain Syndrome (CRPS) is a chronic pain condition that can present specific difficulties when occurring in adolescence. There is limited work exploring future narratives of healthy adolescents, and how these may differ for those who have chronic health conditions, but there is no research on the future narratives of adolescents who have CRPS. **Methods:** In this study, 50 adolescents (44 females, 5 males, 1 preferred not to say) aged 14-25 years (mean=19.8, SD=3.68), completed an online story completion task, with a further sample of 10 completing a follow-up telephone interview. **Results:** Story completion data were initially analysed deductively based on the work of Morley and colleagues using hoped-for and feared-for future codes, revealing higher instances of hope (291 over 48 stories) than fear (99 over 27 stories). These codes were subsequently analysed alongside the in-depth interview data using inductive thematic analysis, generating two themes which represent distinct, yet related, approaches of how adolescents incorporate CRPS into their future narratives: (1) The *centrality of loss* theme identifies how some adolescents described how CRPS brings loss, with narratives focused on how these adolescents imagine such losses continuing into the future, and (2) the *adjusting to loss* theme illustrates how other adolescents were able to imagine a future in which they were able to adjust to the losses which CRPS may bring. **Discussion:** CRPS may damage the future plans of adolescents. However, being or learning how to be flexible about these goals, may help them to build more positive future narratives.

Keywords: chronic pain, adolescence, complex regional pain syndrome, future, qualitative.

Introduction

Adolescence has been recently conceptualised as spanning the 10-25 year age range¹, and is characterised by extensive physical, psychological, and social changes². Identity development, autonomy, and stable peer relationships are key developmental tasks of adolescence³⁻⁵. In addition to these normative developmental tasks, many adolescents face challenges posed by managing chronic pain or long-term health conditions, many of which also include chronic pain⁶. An episode of significant pain, either persistent or intermittent over a three-month period, occurs in up to 38% of youth aged 5 to 18 years with rates increasing over the period of adolescence⁷. Such pain may be related to a disease and injury, alternatively it may be idiopathic in origin⁸. Complex Regional Pain Syndrome (CRPS) is a rare pain condition, with no single identifiable cause⁹. Adolescent onset is common⁹, and related to higher pain-related fear, distress, and functional disability as compared to other pain conditions in this age range^{10,11}.

Adolescents with chronic pain report elevated levels of anxiety, depression, impaired social functioning and pain-related disability¹²⁻¹⁴, with such difficulties continuing into adulthood^{15,16}. Research adopting a developmental approach to the investigation of adolescent chronic pain has demonstrated self-reported delays in education and independence, and enhancement in problem solving¹⁷. Despite the lack of research on identity development in adolescents with chronic pain, there is evidence that fears around how pain will impact one's future are common and increase with age^{18,19}. A more recent study also identified that adolescents with chronic pain perceive pain as negatively impacting how others perceive them, affecting their current and future identity development. However, this study revealed a paradox to pain and development with participants also reporting how in some domains pain enhanced their development²⁰.

Pincus & Morley²¹ explored future thinking and chronic pain in adults. They proposed the Schema Enmeshment Theory to explain the cognitive biases towards pain-related stimuli shown by adults who have chronic pain²¹. This theory suggests that for individuals experiencing chronic pain, their schemata relating to pain, illness, and the self, become increasingly enmeshed over time. Building on this, Morley, Davies & Barton²² asked adults with chronic pain what their hoped-for and feared-for futures looked like, how likely these were to occur, and how dependent on pain they were. The degree to which adults with chronic pain believed their hoped-for and feared-for future selves were dependent on pain, predicted variables such as depressive symptoms, with more dependence related to poorer outcomes²². Such findings suggest that hopes and fears for the future are related to current functioning. Despite adolescence being a critical developmental stage for identity³, including future identity, little is known about how adolescents with CRPS perceive their futures. In this study we build on the work of Morley and colleagues²² to explore the relative frequencies of hoped-for and feared-for future selves, and how these are expressed in the future narratives of adolescents with CRPS. This is achieved through a combination of an online story completion method and subsequent follow-up interviews.

Materials and Methods

Procedure

After ethics review (University of Bath 18-111), participants were recruited to the study via social media platforms (Twitter, Facebook, Instagram, etc.) and CRPS related charity newsletters, blogs, and forums. Recruitment ran for a period of 17 months between May 2018 and September 2019. Interested participants emailed a study specific email address and were asked a series of screening questions prior to being sent a link to the study task. These questions asked when and from whom they received their diagnosis, and were ensure their eligibility

To be eligible to take part in the study, participants were required to be aged 14-25 years with a diagnosis of CRPS. The lower age boundary of 14 was chosen on the basis of pilot work which identified that individuals younger than 14 were unable to manage the task. Participants were also required to have no severe mental health conditions (e.g. psychosis), have the capacity to give informed assent, be fluent in English, and have internet access.

Eligible participants were sent a direct link via email to the online survey platform²³. The online survey platform included information about the study and consent statements. In accordance with UK-based ethical standards, parents/caregivers were required to provide additional consent for participants under 16 years old, prior to the appropriate assent and survey links being sent to the adolescent. This process was automated through the online platform²³ such that the adolescent could not access the survey without the appropriate parental consent in place. Following provision of consent, participants were able to progress to the survey questionnaire screens. These comprised a brief demographics questionnaire and a Numerical Rating Scale (NRS) which asked participants to report current pain using a

typical 0-10 numerical rating scale (ranging from 0 ‘no pain’ to 10 ‘worst possible pain’). This pain intensity rating, along with time since diagnosis, were collected in order to provide a full description of the sample. Participants then proceeded to the story completion tasks where they were asked to write about their own future, their friend’s future, and a previous time when they experienced pain, see below for more details on this method. To enable a rich and detailed exploration of the stories, and to answer the specific research question, we report here only on the stories they wrote about their own futures. After the story completion task, participants were shown a final screen with debrief information and were thanked for their participation. Participants were also asked to provide their email address if they wished to take part in the interview stage of the study. Participants were given a £15 online shopping gift voucher to thank them for their participation. The protocol for the present study is included in the full protocol for a larger set of studies, and was reported in detail and registered on the Open Science Framework²⁴.

Following the 4-point approach described by Robinson²⁵, a total of 43 participants provided details to be contacted for a follow-up interview, of which 10 were selected to give an interview sample who were representative of the overall sample. This selection was based on including a range of participants across demographic categories in addition to participant willingness and availability for interview. Participants provided additional informed consent for the interviews. Interviews were conducted via the telephone, were digitally recorded, transcribed verbatim, and fully anonymised. Participants were given a £10 online shopping gift voucher to thank them for their participation in the interview.

Participants

Stories about their perceived futures were completed by 56 participants. Ineligible (n=6) responses were removed from the dataset, leaving a final sample of 50 participants (44 females; 5 males; 1 prefer not to say). Study participants had a mean age of 19.8 years (range 14-25 years, SD = 3.68), a mean time since CRPS diagnosis of 4.89 years (range <1-11 years, SD = 2.92), and a mean current pain intensity level of 6.1 (range 0-10, SD = 2.15). In depth interviews were conducted with a sub-set of 10 participants (1 male) who completed the story completion task, age range 17 – 25 years (Mean = 21.8, SD = 2.89). These 10 interview participants were self-selected through a question at the end of the story completion task asking participants to indicate if they were interested in taking part in a follow-up interview. Out of those who expressed an interest in the interviews, targeted sampling was used to achieve an interview sample of 10 who were as representative of the story completion sample as possible. See Tables 1, 2 and 3 for further demographic data. The over-representation of females in the story completion and interview samples is similar to that seen in the epidemiology of CRPS populations^{26,27}.

Table 1. *Range, mean and standard deviation for the age, current pain intensity and time since diagnosis of the participants who completed the story completion task and the interviews.*

	Age (years)			Current Pain Intensity			Time Since Diagnosis (years)		
	Mean	SD	Range	Mean	SD	Range	Mean	SD	Range
Story Completion (n=50)	19.8	3.68	14-25	3.1	2.15	0-10	4.89	2.92	<1 - 11
Interviews (n=10)	21.8	2.89	17-25	5.1	2.33	0-9	5.6	3.17	1-11

Table 2. *Gender, education, and employment demographics of the participants who completed the story completion task and the interviews.*

	Gender			Educational Status			Employment Status		
	Male (%)	Female (%)	Unspecified (%)	Full Time (%)	Part Time (%)	None (%)	Full Time (%)	Part Time (%)	None (%)
Story Completion (n=50)	5 (10%)	44 (88%)	1 (2%)	23 (46%)	7 (14%)	20 (40%)	7 (14%)	9 (18%)	34 (68%)
Interviews (n=10)	1 (10%)	9 (90%)	-	4 (40%)	-	6 (60%)	3 (30%)	-	7 (70%)

Table 3. *Country of Residence demographics of the participants who completed the story completion task and the interviews.*

	Country of Residence						
	UK (%)	USA (%)	Australia (%)	Canada (%)	France (%)	Germany (%)	Unspecified (%)
Story Completion (n=50)	32(64%)	9 (18%)	4 (8%)	3 (6%)	1 (2%)	1 (2%)	-
Interviews (n=10)	6 (60%)	-	2 (20%)	-	-	-	2 (20%)

Measures

Story Completion Task

The story completion method²⁸ is a novel technique where participants are provided with a story ‘stem’ and asked to complete the remainder of the story. This particular method allows for efficient collection of larger data sets compared with other more typical qualitative methods²⁹. Story completion studies typically recruit sample sizes of 40-60 participants^{30,31}. Story completion method has typically involved the completion of third-person story stems with no specified time frame for the scenario^{30,32,33}. Due to its focus on the perceptions of adolescents with chronic pain about their future, this study adopted a first-person, future-focused story stem. The stem comprised “*Imagine that it has been 10 years since you graduated from secondary school. You are at your high school reunion and hearing what friends have been doing since you finished school. What will your story look like?*” This story stem was chosen as it was deemed as internationally applicable and set up a social-developmentally relevant scenario. However, to avoid directing participants towards an illness-related narrative, the story stem intentionally did not include reference to CRPS. A minimum length of 900 characters for the future story was set to ensure that data were sufficient for analyses²⁹.

Interviews

All interviews were carried out by ALJ (senior author) and followed a semi-structured interview schedule³⁴. These interviews explored the stories which the participants wrote in more detail, and crucially, asked them about their reasons for choosing this story. This reflection on the stories was key to explore the participants’ understanding of their future stories, how they felt about their future, and how their narrative about their future had changed over time. The interview schedule can be found in the Appendix.

Data Analysis

The data were analysed using a combination of deductive and inductive reflexive thematic analysis^{35,36}. Thematic analysis was selected due to its facilitation of both deductive and inductive analysis. As thematic analysis is theoretically flexible, it is important for us to be clear about the theoretical paradigm we adopted and how this influenced our analysis and interpretation of data. We used the reflexive thematic analysis approach of Braun and Clarke³⁵⁻³⁷, which centres the role of the researcher as an active agent in the analytical process, and requires reflection on the assumptions they bring to their engagement with the data³⁶. Reflexive thematic analysis is widely used in psychology and has been used to analyse both story completion and interview data^{30,32,38}.

Analyses of story completion data

Following the work of Morley and colleagues²² which explored how pain interacted with participants hopes and fears for their future, story completion data were coded for all instances of a ‘hoped for future’ and a ‘feared for future’. Hoped for future codes included statements or partial statements which indicated any element of positivity about the adolescent’s future. Feared for future codes included statements or partial statements which indicated any element of negativity about the adolescent’s future. There was no minimum or maximum length for these statements, but each separate code represented a distinct point. For example, “I will still be in constant and unpredictable pain” was coded as an instance of a feared-for future, and “Hopefully I would be married and maybe a child if I’m able too [sic]” was coded as an instance of a hoped-for future. Table 4 shows these and other examples of the two codes used in the deductive analyses, along with the excerpts they were taken from. Individual stories were first read for familiarisation and then a second time for coding purposes. The hoped-for statements, and feared-for statements were subsequently considered

as two new, independent data sets to be coded inductively using Braun and Clarke's^{36,37} six stage guideline for reflexive thematic analysis.

Table 4: *Examples of deductive coding*

Extract of story	Codes
<i>Hopefully I would be married and maybe a child if I'm able too [sic]. I'd like to be driving by then in a mobility car so I can have my independence.</i> (Becky aged 17, story completion)	Hopefully I would be married and maybe a child if I'm able too [sic] – <u>Hoped-for future</u> I'd like to be driving by then in a mobility car so I can have my independence. – <u>Hoped-for future</u>
<i>I will still be in constant and unpredictable pain which will mean I don't have a job that I am able to keep</i> (Sarah aged 17, story completion)	I will still be in constant and unpredictable pain – <u>Feared-for Future</u> I don't have a job that I am able to keep walk – <u>Feared-for Future</u>
<i>I hope to be graduated from university and in an office job as standing for too long is unbearable.</i> (Sophie aged 18, story completion)	I hope to be graduated from university and in an office job – <u>Hoped-for Future</u> Standing for too long is unbearable – <u>Feared-for Future</u>

Analyses of interview data

Interview data were coded using reflexive inductive thematic analysis^{36,37}. An inductive reflexive approach was adopted to facilitate in-depth analyses of the expanded detail around the stories in addition to reflections around the reasons for selecting particular stories. Such

an approach also facilitated analysis of the researcher's dynamic influence in the conversation with the participant. The interview data were analysed following the coding and initial theme development of the story completion data. This allowed the authors to use the coding of the interview data as a check to confirm that the themes generated from analysis of the story completion data were congruent with participants' understanding of their future plans in the subsequent interview data. This process captured the participant's understanding of their stories.

Combining deductive and inductive analyses

A deductive-inductive two-stage analytical process was adopted to facilitate exploration of varied aspects of the data, since both approaches are distinct analytical approaches which explore different levels of meaning within the data. Deductive analysis is a 'top-down' approach which captures manifest meaning within the data, whereas inductive analysis is a 'bottom up' approach which captures latent meaning³⁹. The combination of both approaches allows for exploration of both levels of meaning, giving a richer and more detailed analysis of the data. However, this detailed level of analyses meant that it was only possible to analyse participants stories of their own futures, rather than the other stories collected as part of the wider study.

Quality in qualitative research

Prior to data analysis, issues related to ensuring high quality and rigour in qualitative research were considered. While we have chosen data collection methods which can provide rich and high-quality data, it is important to be aware of the researchers' key role as the "instrument for analysis" and therefore our responsibility for the quality of the research⁴⁰. Numerous criteria exist which can be used to evaluate the quality and rigour of the analyses including

credibility, transferability, dependability, and confirmability⁴⁰⁻⁴². Below, we have outlined the particular ways in which we have explicitly considered, actively implemented and embedded these principles within our study.

First, we focused on collecting and analysing two forms of data within the study (interview and story completion data) to enable a credibility check on the themes within the analyses. The addition of the interview data helped to confirm that our understanding of the stories was aligned with participants' perceptions and sense making of their stories, thereby increasing the confirmability of the results. Throughout the results section, we have included quotations from the data, including pseudonyms, ages, gender, and whether the quotation was derived from the stories or interviews. These quotations are taken from range of stories and interviews across the participant sample, providing accurate examples of the themes across the whole data. These quotations demonstrate how the themes are grounded in the data, and give the reader a clear understanding of the data and their context. Finally, throughout the data analysis, the authors took a reflexive approach, both individually and collectively as a group, helping us to be aware of our influence on the analysis. AJ (first author) led the data analysis, supported by ALJ and LC. This subset of authors had many group discussions and feedback at all stages of theme development, and subsequently all authors participated in such discussions throughout theme finalisation and manuscript write up. These discussions ensured that author interpretations and inputs into theme development were credible and grounded in the data.

Results

Deductive analyses

The deductive analysis generated 291 instances of hoped-for futures across 49 of the 50 stories, and 99 instances of feared-for futures across 27 of the 50 stories. Only one story contained exclusively feared-for codes and 23 contained exclusively hoped-for codes. Just over half of the stories ($n = 26$) included instances of both hoped-for and feared-for future, with some stories containing both codes within the same sentence, for example: *“At this point it has proven to be increasingly more difficult to see my dreams as easily achievable while this will not stop me it definitely makes my dreams seem unachievable”* (Megan, 21, story completion). These frequencies highlight that hoped-for statements dominated participants’ stories.

Inductive analyses

To extend our understanding of adolescents’ perceptions of their future selves, the hoped for ($n = 291$) and feared for codes ($n = 99$) from the story completion task were used to generate initial themes, before being combined with the interview data. We explored the different narratives that the participants created regarding their perceived future selves and the role that CRPS may play in these futures. Overall, the stories suggested that prior to the onset of CRPS, participants perceived that they would have freedom to choose their future path, and that their lives would involve meeting a set of recognised developmentally normative milestones (e.g. educational and employment achievements, and starting a family). The onset of CRPS was associated with losses, to a greater or lesser extent, around these expected futures. The reflexive thematic analysis generated two themes: *Centrality of Loss*, and *Adjusting to Loss*, which are explored in detail below with quotations from both the stories

and interviews. These themes reflect differences in how the participant narratives frame the losses associated with CRPS, and crucially, how they describe their expectations around continued loss or adjustment to these losses into their future.

Centrality of Loss

The theme of centrality of loss captures the idea that participants described stories which centred around the losses they perceived in their future due to CRPS. The exact nature and severity of this loss varied, but such stories described how the adolescent's desired future had been taken from them as a result of developing and living with CRPS. Such losses were a central focus these narratives.

For some participants, living with CRPS was perceived to be incompatible with their future plans, reflecting a broad sense of loss concerning their imagined future and its possibilities. For instance, Amy (aged 24) described in her story how CRPS had destroyed her plans for the future, leaving only a void and a memory of what she had once had. She described a sense of loss and hopelessness, and appeared to be grieving for the life she feels she had lost. *"I [would] talk about what I had, not what I have. I [would] talk about my achievements but not my hopes and dreams. I hope no one wants to keep in touch because I don't have much of a life anymore."* Amy continues to describe how the *"future only looks bleak."* For participants such as Amy, CRPS is an all-consuming force which insidiously creeps into their futures, denying these young people the opportunities and experiences that they rightfully expected would feature in their imagined futures. Participants also talked about the losses they are currently experiencing due to CRPS, and how they believed the CRPS, and its impact on their life, will deteriorate over time in a trajectory of continued loss. As an exemplar of this, Katy (aged 25) describes in her story how *"I'll probably have declined in*

health, mobility will be less, pain worse, muscle wastage more noticeable and quality of life will have declined more (even though right now it feels hard to see how things could get worse)”. Extending this further, Natasha (aged 19) described in her interview “I think it’s a condition that changes who you are” and “I just completely lost myself, I’m not anywhere near the person that I was.” Here, Natasha shows how deep and enduring the loss she has experienced due to CRPS is. She is expressing how CRPS has changed her identity, leading to a loss of her identity and a loss of control over herself, which she believes will persist into her future. In contrast, other stories described losses which were less extreme, yet remained central in their future narratives. For instance, Hannah (aged 19) described in her story how she felt she had lost the ability to work full time due to the CRPS: “I don’t think I’d be able to handle full time because of bad nights and flare ups”. Although Hannah expresses that she believes that she will not lose the ability to work altogether, her focus is on what she has lost rather than what she has retained.

Loss narratives were viewed through a social lens, in which participants engaged in an active process of comparing their imagined futures with those of their peers, and portrayed overly negative social responses. For example, Lisa (aged 25) described how she will *“have to explain that I don’t work. With that I would have to explain why and I would feel ashamed.”* Lisa’s shame is compounded by the comparison with their peers, who are assumed to be able to work without problems. The following quotation from Louise’s (aged 17) story demonstrates how the participants’ perceptions of the way others see them, can amplify this sense of loss and negative feelings they have about themselves. *“People may have given up on me as they think I can’t be on crutches for that long and therefore believe I’m faking or it’s psychosomatic and this leads to loss of friends and further social isolation.”* How far Louise’s perception is supported by others’ views is not known.

In addition to the direct comparison between what they and their peers will achieve, another important element to the social context of the sense of loss was the loss of shared experience with their peers, and the isolation this brings. For example, Sarah (aged 17) described in her story: *“I probably won't have many friends because they will all be off building their futures, knowing where they want to go in life, whilst I'm at home alone wondering if I will ever have a pain free day again”*. For Sarah, there seemed to be an inability to build a meaningful future and this creates an increasing distance between her and her peers in terms of possible futures, highlighting a further sense of loss. For others, this sense of social loss was focused on the dominance of CRPS in their life. Lisa (aged 25) describes in her narrative how *“I won't have much to talk about other than pain”*, highlighting her sense of being increasingly consumed by CRPS, and the loss of non-pain elements of herself and her life. For Lisa, the sense of loss centres on a narrowing of her experiences, extending the perceived gap between Lisa and her peers as there is no common ground in terms of shared experiences and imagined future goals.

Adjusting to Loss

Many participants described their future goals as achievable despite living with CRPS and the losses it may bring. In such instances, participants' narratives did not have loss as a central component. Rather, they described how they will have adjusted to such losses, and how their future plans have been adapted to accommodate living with CRPS in a positive way.

Some participants described an expectation of achieving the goals they had prior to CRPS, such as marriage, families and careers, with some not even mentioning CRPS at all when describing their imagined future. For example, Jo (aged 17) explained in her story how she

will “*become a paramedic or ICU Pediatric nurse after attending university*”, with Ali (aged 21) saying in her story “*I will hopefully be married and have children*”. Other stories mentioned CRPS but explicitly stated that it will improve substantially. For example, Andrew (aged 14) stated in his story “*I will have little or no chronic pain*”, and Sam (aged 15) said “*I will be able to live CRPS free and not be at risk of having a flare up every time I fall over*”. Such narratives show that some participants do not envisage any long-term loss of functioning due to CRPS. Contrastingly, other participants do foresee some losses due to CRPS in their future, but focus on describing how they imagine that their future will include a positive trajectory despite the current losses due to CRPS. This positive trajectory may be due to various reasons, such as a perceived reduction in the severity of CRPS over time. Jo (aged 17) expressed such a perception in her interview: “*my flare ups will continue but they’re not gonna be as intense or as often as they are now*”. For others, their stories suggest a perception that they will adjust to the losses that CRPS brings, and be more skilled in managing the condition. Vicky (aged 19) stated this in a general sense in her story: “*I will be able to cope better and not let it overtake my life*”. Laura (aged 24) described in her story the details of how she will achieve this: “*I’d like to think that by this point I’d be on the right medication and have a better grasp on what causes my triggers and what to avoid. I also hope I will know the right ways to calm myself down after an attack or episode so it doesn’t eat up as much time as it used to.*” Such expectations of abating symptoms or improved self-management strategies may explain why there is such an abundance of hope present in these stories. However, as described below, a number of participants displayed hope despite an expectation that CRPS symptoms would continue to have a substantial impact on their future.

Several stories outlined that for some, CRPS had created more substantial losses and deviation from their original future plans. However, in contrast to those in the sense of loss

theme, these participants presented the changes in an adjustment-focused rather than a loss-focused narrative. For example, Clare (aged 18) described in her story how she imagines that she will be *“living independently with potentially a service animal to aide me in things I can’t do on my own”*. When discussing her future story in the interview, Ellie (aged 23) described her perceived need to change her career path to one that may better meet restrictions associated with her pain condition: *“I had to adapt from that sort of, the story I had for myself. But it’s essentially the same thing that I’ve wanted for myself since I was about ten. Just with slight adaptations to allow for the difficulties that CRPS brings in. So, like rather than sort of being Ed Psych, going into counselling instead?”*. Some participants extended this further and perceived the experience of living with CRPS as offering them unique opportunities for future direction and societal integration that would otherwise not be possible. These participants provided narratives where their current struggles would be constructively used to carve out a positive future that enables them to use the skills they had gained from managing a complex pain condition. For example, Jo (aged 17) described in her story how she will be *“be working as a volunteer to help raise money for treatment of CRPS and new research into the care plans needed”*. For this participant, her experiences would enable her not only to have a meaningful career, but to advance scientific knowledge in the area of CRPS. Stories such as Jo’s suggest that over time, the participant’s future will be dominated by CRPS in a positive way, enabling them to turn negative experiences into positive ones, both for themselves and others. This may represent the participants’ pursuit of meaning, which their previous plans are no longer able to provide and so they create new plans which would allow them to find meaning out of their experiences of CRPS. Such narratives suggest an active process of engaging with the loss, and using the experience as a catalyst for personal growth and transformation. Rachel (aged 18) summed this up when she

wrote in her story that *“I hope to have turned this ugly situation into something more beautiful.”*

Achieving success in their imagined future, despite potential current or future losses due to CRPS, was important for many participants, and as described above, this ranged from achieving their pre-diagnosis goals, to the creation of entirely new aspirations and goals.

Such success was often described at an individual level (e.g. a lucrative and professionally engaging career), however, for some participants, the social context in which this success is perceived was critical. For Debbie (aged 25, quotation from her story), it was critical that

“People would be surprised to see the things I’ve accomplished, and happy to see my success.” Similarly, Rachel (aged 18) stated in her story, *“I want to say to all the kids in high school who told me I couldn’t: ‘look what I did... all the things you said I would never’.”*

These quotations suggest that while Debbie and Rachel imagine a future where they are able to achieve their goals despite the losses associated with CRPS, they are aware of the perceptions of their peers. Debbie and Rachel express a belief that their peers’ perceptions of them are of continued loss and inability to achieve. Success was not sufficient for some participants, with Jo (aged 17) describing in her story how *“My secondary school friends will be in awe that despite my numerous obstacles and conditions that I still managed to achieve my dream”*.

Discussion

We explored how adolescents with CRPS think about, and imagine, their futures. Study findings identified the dominance of loss in young people's narratives in relation to CRPS, both with regard to the impact on their lives and the active role of adjustment to such losses. Themes comprised 'centrality of loss' and 'adjusting to loss'. In particular, the centrality of loss theme illustrates how for some adolescents with CRPS, their perceptions of the future are dominated by the losses associated with their condition. Contrastingly, the adjusting to loss theme shows that other adolescents are able to adapt and adjust their future plans to overcome such losses.

Adolescence is a time of change and development, when individuals start to consider possible futures and how they will build such futures independently of their parents and other caregivers. For individuals who experience CRPS during adolescence, loss (or perceived imagined loss) of some, or all, of their future plans becomes a consideration. Such findings are congruent with the wider chronic illness literature in which ill-health has been shown to disrupt individual's lives and challenge "...earlier taken-for-granted assumptions"⁴³. Such disruption necessitates some degree of narrative change to accommodate the new demands of managing a chronic illness⁴⁴. In terms of the specific narrative changes which occur in youth with chronic illnesses, research suggests that the additional difficulties that may come with having such an illness may impact one's hopes for the future. Additionally, there may be a unique impact of developing an illness, rather than simply having an illness, which increases this sense of loss⁴⁵. This is consistent with the findings of Stommen and colleagues¹⁸, who found that adolescents with chronic pain perceived more barriers to achieving future goals as compared to pain-free adolescents. Situating our findings within this chronic illness literature, it is possible that when adolescents face challenges associated with managing CRPS, they

may engage in a process of re-evaluating their identity and future trajectory. Consequently, although adolescents may initially present as being confronted by loss it is possible that over time they are able to adapt to such losses. Initial theme presentation, and subsequent movement towards a more loss-focused or adjustment-focused narrative may be facilitated by individual factors such as the extent of disability, co-occurring mental illness, and duration of CRPS. Additionally, factors external to the person such as parental mental illness, social support, journey through health services, and therapeutic intervention, may also facilitate such change.

The analysis for this study was informed by the work of Morley and colleagues²², who asked adults with chronic pain about their hopes and fears for their future. The results of the present study suggest that such a theoretical approach may also be applicable to adolescents. Our participants expressed much more hope than fear. These were often combined and presented in narratives which had loss or adjustment as their focal point, suggesting a complex interaction between loss, fear, hope, and adjustment. Carver's⁴⁶ theoretical model of loss adjustment, which proposes 4 potential trajectories of functioning following initial losses in response to adverse events, may help us to untangle this complex relationship. Carver suggests that individuals may 1) continue on a negative loss trajectory, 2) partially recover some functioning, 3) return to prior functioning (which Carver defines as resilience), or 4) may surpass prior functioning (which Carver defines as thriving). Carver describes how resilience and thriving may only occur when a negative event is appraised as being a challenge which includes the potential for growth, rather than a threat. Applying this model to our results, we can see all four of these trajectories. The narratives expressed in the centrality of loss theme appear to be continuing on a loss trajectory as they are currently unable to adjust to the losses that CRPS has imposed on them. The three other trajectories from

Carver's model nicely illustrate the variety of stories observed within our adjusting to loss theme, with some participants accepting limitations on their future functioning, some expecting limited or no impact on their functioning, and others take something positive from their CRPS experience which they otherwise would not have. Carver's model suggests that these different trajectories may be related to the degree to which participants appraise CRPS as a threat or a challenge. Previous research which applied Carver's model to adults with arthritis found that many people were thriving but only after several years following diagnosis, suggesting an initial sense of loss, and the need for time to adapt⁴⁷. This study was cross-sectional, and only explored the trajectories which the participants imagine they will have, from the perspective of a single time-point. Therefore, a longitudinal investigation is needed to explore the real-life trajectories of adolescents with CRPS, how their future narratives may change over time, and the continued relationship between the two. Also, previous research has shown a combination of functional enhancement and reduction depending on the area of functioning¹⁷. Therefore, an overall model of functional trajectories may be too simplistic to capture the complexities of loss and loss adjustment shown in the present study. It may be more accurate to consider functioning as a multidimensional construct, where individuals may have numerous different trajectories. A multidimensional model of functioning may be useful when considering adolescent development in general. However, it may be particularly relevant when considering the development of those with chronic illnesses such as CRPS. Such an approach could help to identify in what specific areas an individual is experiencing difficulties, and where they may be excelling. This would therefore help to target interventions, and highlight areas of success which may be overlooked. Future research is needed to explore adjustment trajectories and the mechanisms underlying them. Specifically, an exploration is needed of factors such as challenge and threat appraisals, which may drive differences in adjustment trajectories.

This study of course includes some strengths and limitations. A particular strength of the study is the use of a novel method, story completion, to explore participant's future narratives, an approach previously not taken in the context of paediatric chronic pain. Combining this with detailed interviews resulted in a wealth of rich data, while the use of both deductive and inductive thematic analysis yielded robust results. With regard to limitations, our study findings are limited to the experiences of this particular sample of young people with CRPS, and consequently, it is not possible to draw comparisons with young people without CRPS or with other long-term conditions. Second, data were collected at a single time point, limiting analyses to those at such a single time point and preventing us from extrapolating to the stability of the observed perceptions over time. Third, we used a story stem which described a school reunion as it is a situation which most participants could related to, and where they would be telling people about their lives. However, it may have led some participants to write stories which were more socially orientated than they would have otherwise written. Finally, we did not ask participants about their treatment history and consequently, it is unclear how far adjustment to loss has been therapeutically mediated or arrived at independently.

Overall, this study provides valuable insights into the future narratives of adolescents who have CRPS. Such individuals appear to be overall hopeful about their future, although this hope is interspersed with concerns about how CRPS may impact their lives. Adolescents who have CRPS may see their condition as an insidious force which results in a variety of losses in their life. For some adolescents, their future narratives centre around these losses which they imagine will only deepen as they move through life. For others, their future narratives

are focused on their ability to build a positive and fruitful future through adaptation to such losses.

References

1. Sawyer SM, Azzopardi PS, Wickremarathne D, Patton GC. The age of adolescence. *Lancet Child Adolesc Heal*. 2018;2(3):223-228. doi:10.1016/S2352-4642(18)30022-1
2. Christie D, Viner R. ABC of Adolescence: Adolescent development. *BMJ*. 2005. doi:10.1136/bmj.330.7486.301
3. Schwartz SJ, Petrova M. Fostering healthy identity development in adolescence. *Nat Hum Behav*. 2018. doi:10.1038/s41562-017-0283-2
4. Pavlova MK, Haase CM, Silbereisen RK. Early, on-time, and late behavioural autonomy in adolescence: Psychosocial correlates in young and middle adulthood. *J Adolesc*. 2011. doi:10.1016/j.adolescence.2010.04.002
5. Poulin F, Chan A. Friendship stability and change in childhood and adolescence. *Dev Rev*. 2010. doi:10.1016/j.dr.2009.01.001
6. O'Donohue W, Tolle LW. Introduction: Adolescents with Chronic Illnesses: Issues and Answers. In: O'Donohue W, Tolle LW, eds. *Behavioral Approaches to Chronic Disease in Adolescence: A Guide to Integrative Care*. London: Springer; 2009:3-6.
7. King S, Chambers CT, Huguet A, et al. The epidemiology of chronic pain in children and adolescents revisited: A systematic review. *Pain*. 2011;152(12):2729-2738. doi:10.1016/j.pain.2011.07.016
8. Malleson PN, Connell H, Bennett SM, Eccleston C. Chronic musculoskeletal and other idiopathic pain syndromes. *Arch Dis Child*. 2001;84(3):189-192. doi:10.1136/adc.84.3.189
9. Weissmann R, Uziel Y. Pediatric complex regional pain syndrome: a review. *Pediatr Rheumatol*. 2016;14.
10. Simons LE. Fear of pain in children and adolescents with neuropathic pain and complex regional pain syndrome. In: *Pain*. Vol 157. Lippincott Williams and Wilkins;

2016:S90-S97. doi:10.1097/j.pain.0000000000000377

11. Logan DE, Williams SE, Carullo VP, Claar RL, Bruehl S, Berde CB. Children and adolescents with complex regional pain syndrome: More psychologically distressed than other children in pain? *Pain Res Manag*. 2013;18(2):87-93.
doi:10.1155/2013/964352
12. Asmundson GJG, Noel M, Petter M, Parkerson HA. Pediatric fear-avoidance model of chronic pain: Foundation, application and future directions. *Pain Res Manag*. 2012.
doi:10.1155/2012/908061
13. Noel M, Groenewald CB, Beals-Erickson SE, Gebert JT, Palermo TM. Chronic pain in adolescence and internalizing mental health disorders: A nationally representative study. *Pain*. 2016;157(6):1333-1338. doi:10.1097/j.pain.0000000000000522
14. Forgeron PA, King S, Stinson JN, Mcgrath PJ, Macdonald AJ, Chambers CT. Social functioning and peer relationships in children and adolescents with chronic pain: A systematic review. *Pain Res Manag*. 2010;15(1):27-41.
15. Shelby GD, Shirkey KC, Sherman AL, et al. Functional abdominal pain in childhood and long-term vulnerability to anxiety disorders. *Pediatrics*. 2013;132(3):475-482.
doi:10.1542/peds.2012-2191
16. Walker LS, Sherman AL, Bruehl S, Garber J, Smith CA. Functional abdominal pain patient subtypes in childhood predict functional gastrointestinal disorders with chronic pain and psychiatric comorbidities in adolescence and adulthood. *Pain*. 2012.
doi:10.1016/j.pain.2012.03.026
17. Eccleston C, Wastell S, Crombez G, Jordan A. Adolescent social development and chronic pain. *Eur J Pain*. 2008;12(6):765-774. doi:10.1016/j.ejpain.2007.11.002
18. Stommen NC, Verbunt JA, Goossens ME. Future goals of adolescents and young adults with chronic musculoskeletal pain. *Eur J Pain (United Kingdom)*.

- 2015;20(4):564-572. doi:10.1002/ejp.762
19. Meldrum ML, Tsao JCI, Zeltzer LK. "I Can't Be What I Want to Be": Children's Narratives of Chronic Pain Experiences and Treatment Outcomes. *Pain Med.* 2009;10(6):1018-1034. doi:10.1111/j.1526-4637.2009.00650.x
 20. Jordan A, Noel M, Caes L, Connell H, Gauntlett-Gilbert J. A developmental arrest? Interruption and identity in adolescent chronic pain. *Pain Reports.* 2018;3(7). doi:10.1097/PR9.0000000000000678
 21. Pincus T, Morley S. Cognitive-processing bias in chronic pain: A review and integration. *Psychol Bull.* 2001;127(5):599-617. doi:10.1037/0033-2909.127.5.599
 22. Morley S, Davies C, Barton S. Possible selves in chronic pain: Self-pain enmeshment, adjustment and acceptance. *Pain.* 2005;115(1-2):84-94. doi:10.1016/j.pain.2005.02.021
 23. Qualtrics [Online Computer Software]. 2019. <https://www.qualtrics.com>.
 24. Rugg T, Caes L, Malcolmson L, et al. Telling their story: Exploring future perceptions in adolescents with Complex Regional Pain Syndrome and their parents/caregivers. A protocol. *Open Sci Framew.* 2018. <https://osf.io/r2wsc/>.
 25. Robinson OC. Sampling in Interview-Based Qualitative Research: A Theoretical and Practical Guide. *Qual Res Psychol.* 2014;11(1):25-41. doi:10.1080/14780887.2013.801543
 26. Tan ECTH, Zijlstra B, Essink ML, Goris RJA, Severijnen RSVM. Complex regional pain syndrome type I in children. *Acta Paediatr Int J Paediatr.* 2008;97(7):875-879. doi:10.1111/j.1651-2227.2008.00744.x
 27. Low AK, Ward K, Wines AP. Pediatric Complex Regional Pain Syndrome. *J Pediatr Orthop.* 2007;27:567-572.
 28. Clarke V, Braun V, Frith H, Moller N. Editorial Introduction to the Special Issue:

- Using Story Completion Methods in Qualitative Research. Qual Res Psychol.* 2019;0(00):1-20. doi:10.1080/14780887.2018.1536378
29. Clarke V, Hayfield N, Moller N, Tischner I. Once Upon a Time... Qualitative Story Completion Methods. In: Braun V, Clarke V, Gray D, eds. *Collecting Qualitative Data: A Practical Guide to Textual, Media, and Virtual Techniques*. Cambridge: Cambridge University Press; 2017:45-70.
 30. Clarke V, Braun V, Wooles K. Thou shalt not covet another man? Exploring constructions of same-sex and different-sex infidelity using story completion. *J Community Appl Soc Psychol.* 2015. doi:10.1002/casp.2204
 31. Hayfield N, Wood M. Looking heteronormatively good! Combining story completion with *Bitstrips* to explore understandings of sexuality and appearance. *Qual Res Psychol.* 2018;00(00):1-21. doi:10.1080/14780887.2018.1536390
 32. Frith H. Accounting for orgasmic absence: Exploring heterosex using the story completion method. *Psychol Sex.* 2013. doi:10.1080/19419899.2012.760172
 33. Walsh E, Malson H. Discursive constructions of eating disorders: A story completion task. *Fem Psychol.* 2010. doi:10.1177/0959353509350759
 34. Mason J. Semi structured interview. In: Lewis-Beck MS, Bryman A, Liao TF, eds. *Encyclopedia of Social Science Research Methods*. Thousand Oaks, California: SAGE Publications Inc.; 2004:1021-1022.
 35. Braun V, Clarke V. *Successful Qualitative Research*. London: SAGE Publications Inc.; 2013.
 36. Braun V, Clarke V. Reflecting on reflexive thematic analysis. *Qual Res Sport Exerc Heal.* 2019;11(4):1-9. doi:10.1080/2159676X.2019.1628806
 37. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol.* 2006. doi:10.1191/1478088706qp063oa

38. Holmqvist K, Frisén A. "I bet they aren't that perfect in reality:" Appearance ideals viewed from the perspective of adolescents with a positive body image. *Body Image*. 2012;9(3):388-395. doi:10.1016/j.bodyim.2012.03.007
39. Clarke V, Braun V. Thematic analysis. *J Posit Psychol*. 2017;12(3):297-298. doi:10.1080/17439760.2016.1262613
40. Nowell LS, Norris JM, White DE, Moules NJ. Thematic Analysis: Striving to Meet the Trustworthiness Criteria. *Int J Qual Methods*. 2017;16(1). doi:10.1177/1609406917733847
41. Hannes K. Critical appraisal of qualitative research. In: Noyes J, Booth A, Hannes K, et al., eds. *Supplementary Guidance for Inclusion of Qualitative Research in Cochrane Systematic Reviews of Interventions. Version 1*. Cochrane Collaboration Qualitative Methods Group; 2011.
42. Shenton AK. *Strategies for Ensuring Trustworthiness in Qualitative Research Projects*. Vol 22. IOS Press; 2004.
43. Charmaz K. *The Body, Identity, and Self: Adapting to Impairment*. Vol 36.; 1995.
44. Frank A. *The Wounded Storyteller: Body, Illness and Ethics*. Chicago: University of Chicago Press; 1995.
45. Venning AJ, Elliott J, Whitford H, Honnor J. The impact of a child's chronic illness on hopeful thinking in children and parents. *J Soc Clin Psychol*. 2007;26(6):708-727. doi:10.1521/jscp.2007.26.6.708
46. Carver CS. Resilience and thriving: Issues, models, and linkages. *J Soc Issues*. 1998;54(2):245-266. doi:10.1111/0022-4537.641998064
47. Sirois FM, Hirsch JK. A longitudinal study of the profiles of psychological thriving, resilience, and loss in people with inflammatory bowel disease. *Br J Health Psychol*. 2017;22(4):920-939. doi:10.1111/bjhp.12262

Appendix – Interview Schedule

Topic	Question and Prompts
Ice Breaker & Background	Can you please tell me about your pain condition?
Future Self Story	<p>I would like to know more about the first story completion task you did, about your own future story. Please can you tell me about your future story?</p> <p><i>Prompts: How will this look? Can you tell me what you think you will be doing work/study wise? Can you tell me about the relationships you will have? What do they look like? Who are they with? Friends, family, romantic partners? Where do you think you will be living, and with who?</i></p> <p>What are your reasons for choosing this story for yourself?</p>
Friend's Future Story	<p>Now I would like to talk about the second story completion task that asked you about your friend's future. Please can you tell me about what you think your average friend's future story will be?</p> <p><i>Prompts: How will this look? Can you tell me what you think they will be doing work/study wise? Can you tell me about the relationships they will have? What do they look like? Who are they with? Friends, family, romantic partners? Where do you think they will be living, and with who?</i></p>

	What are your reasons for choosing this story for your friends?
Both Stories	<p>Can you tell me how the two stories compare?</p> <p><i>Prompts: Please can you tell me why you think you and your friend's stories will be different/similar?</i></p>
General Thoughts About Pain and Future	<p>Can you tell me about anything else that you think we should know about in terms of living with pain and thinking about your future?</p>